YOUSSEF SYNDROME REVISITED

Aruna Menon[1], Prajakta S. Mehendale[2], Srinivas Sangisapu[3], Ajai Srivastava[4], Shilpa Asthana[5]

[1]Professor Indian Army, Department of Obstetrics and Gynaecology, INHS, Asvini, Mumbai, India.
[2]Gynaecologist, Department of Obstetrics and Gynaecology, INHS, Asvini, Mumbai, India.
[3]Classified Specialist, Department of Obstetrics and Gynaecology, INHS, Asvini, Mumbai, India.
[4]Senior Advisor, Department of Obstetrics and Gynaecology, MH, Devlali.
[5]Classified Specialist, Department of Obstetrics and Gynaecology, INHS, Asvini, Mumbai, India.

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PRESENTATION OF CASE

Youssef's syndrome, a rare entity, consists of cyclical haematuria or menouria, amenorrhea associated with vaginal leakage of urine. This was first described by M Youssef, an Egyptian Surgeon in 1957.[1] Since then, reports of this rare urogenital fistula have appeared off and on in literature. We present a case subsequent to a ruptured uterus during a trial of labour after a previous Caesarean section.

A 26-year-old lady, para 2, presented to the OPD with complaints of intermittent haematuria, increased frequency of micturition and lactational amenorrhea. She also gave history of occasional watery discharge per vaginum, which had subsided by the time of presentation.

Her obstetric history revealed that she had 2 live children. The first pregnancy resulted in a caesarean section for breech presentation. During the subsequent pregnancy, she underwent an induction of labour for VBAC in a teaching hospital. This unfortunately resulted in a ruptured uterus following administration of dinoprostone gel and augmentation with oxytocin. A live male baby was delivered by emergency LSCS. The ruptured uterus was cruciate in nature extending up to the bladder and was repaired with Vicryl followed by tubectomy. No injury to the bladder was noticed at that time. She was treated with antibiotics and kept catheterised for 7 days. She had an uneventful hospital stay subsequently and was discharged after 10 days.

DIFFERENTIAL DIAGNOSES

1. Urinary tract infection.
2. Vesicouterine fistula.

At the time of presentation, she was found to have features suggestive of UTI, which was treated with antibiotics.

As the symptoms persisted, the patient was taken up for a cystoscopy examination. This revealed a rent in the dome of the bladder through, which blood appeared to be streaming into the bladder with a few clots adherent to the point of entry.

A possibility of vesicouterine fistula was entertained and a hysterosalpingogram and an intravenous pyelogram were done. These investigations, however, proved noncontributory and inconclusive. She then underwent an office hysteroscopy. The hysteroscopy confirmed the presence of a vesicouterine fistula as the catheter placed in the bladder could be visualised through the hysteroscope in the uterus. However, as the uterine cavity could not be distended due to the fistula, hysteroscopy was abandoned.

CLINICAL DIAGNOSIS

Vesicouterine fistula.

PATHOLOGICAL DISCUSSION

A vesicouterine fistula was an obvious and constant source of infection to the urinary tract and needed to be treated surgically in this case.

DISCUSSION OF MANAGEMENT

The patient was then taken up for an exploratory laparotomy. The uterine rent was repaired. The urinary bladder was opened sagittaly, the fistulous tract identified and excised. The bladder was repaired in 2 layers after placing a suprapubic catheter through a cystostomy and an indwelling Foleys catheter per urethra. A patch of omentum was interposed between the bladder and uterus. An intraabdominal drain was placed before closure. The postop recovery was uneventful and the patient was discharged with advice to take a continuous preparation of oestrogen-progesterone in the form of oral contraceptive pills for a period of 3 months.

Discussion of Literature

It was in 1957, that AF Youssef, an Egyptian surgeon first described a condition where a patient presented with amenorrhea, menouria (a term coined by Youssef to describe the passage of menstrual blood through the urethra following a vesicouterine fistula) and vaginal leakage of urine.[1]

The causes are usually secondary to caesarean section, abnormal or instrumental delivery and rupture of the bladder and uterus.[2] Anecdotal cases are also reported due to other causes like myomectomy, insertion of IUCD, rupture of bladder and uterus due to trauma. In the index case too, the cause is attributable to a previous CS scar.

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Corresponding Author:
Dr. Prajakta S. Mehendale,
Gynaecologist, Department of Obstetrics and Gynaecology,
INHS, Asvini, Mumbai, India.
E-mail: prajdoc@gmail.com, aruna.menon@gmail.com
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Although, uncommon, bladder injury during caesarean delivery can be associated with significant morbidity. Potential complications may include prolonged operative time, urinary tract infection, prolonged indwelling catheter time and formation of vesicouterine or vesicovaginal fistula. The reported incidence of bladder injury with cesarean delivery ranges from 0.14% to 0.94%. The largest series of 23 bladder injuries reported by Eisenkop et al in 1982 demonstrated an overall incidence of 0.31% with an incidence of 0.19% in primary caesarean deliveries and 0.6% in repeat caesarean deliveries.[3]

The complication can occur in the immediate postop period, puerperium or rarely even later.

The investigative modalities include cystoscopy, HSG, intravenous pyelogram, sonosalpingogram and hysteroscopy.[4] A recent report mentions MRI as the most useful tool of diagnosis wherein fluid is demonstrated within the fistula obviating the need for conventional radiographic contrast examination. The advantage of an IVU is that it may delineate an associated ureteric injury. In our case, an IVU and HSG proved inconclusive, probably because lateral view films were not taken. However, cystoscopy and hysteroscopy clinched the diagnosis. No further sophisticated investigations suggested in the foregoing paragraph were found necessary.

The treatment options are both medical and surgical. Spontaneous healing is reported in 5% of cases.[5] The conservative management by only catheterising the patient for a period of 10-14 days can be tried.[6] The other medical modality tried is hormonal where the patient is rendered amenorrheic using oestrogens, thereby promoting healing and preventing the shedding of endometrium. 41 such cases have been reported so far.[7] As the rent in the case being reported was large, no attempt was made to try out this modality of treatment.

The mainstay of treatment remains surgical- the route may be abdominal and the surgery will depend on the reproductive history and desire of the patient for further reproductive capacity.[8]

Patients scheduled for surgery should undergo pre-treatment of urinary tract infections. Surgical repair of vesico-uterine fistulas is performed by different approaches which include the vaginal, transvesical-retroperitoneal and transperitoneal access, the last of which is considered the most effective with the lowest relapse rate. Recently, laparoscopy has been proposed as a valid option for repairing vesicouterine fistulas. The endoscopic treatment may be effective in treating small vesicouterine fistulas.[5]

The pregnancy rate after repair is 31.25% with a rate of term deliveries of 25%.[5]

It is advisable that after vesicouterine fistula repair delivery should be performed by repeating a caesarean section to avoid the risk of fistula recurrence. 95% of patients with vesicouterine fistula will undergo another operation for repairing the fistula. In the meantime, they are bothered by related symptoms, which impair their quality of life.[9] Intraoperative diagnosis is the gold standard in detecting vesicouterine fistulas for allowing immediate repair. Intraoperative sonography by the transvaginal route for haematuria is useful when suspecting bladder injury, while dissecting the uterine lower segment.[6,9] This may allow a prompt repair of the iatrogenic fistula.

This case is reported to highlight the pitfalls of improper closure and subsequent implications and complications for the patient.

Figure 1. Cystoscopy YSF

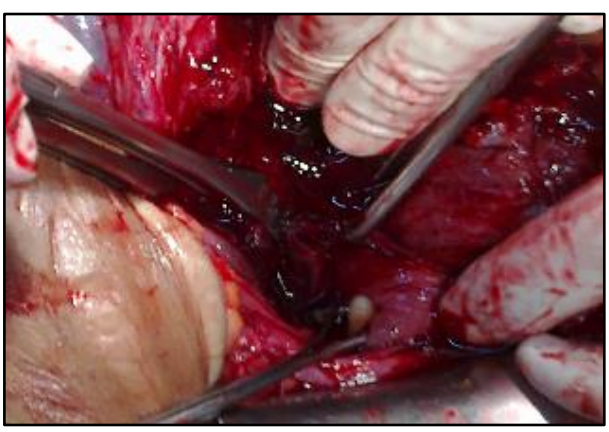

Figure 2. Picture of Laparotomy

FINAL DIAGNOSIS

Vesicouterine Fistula.

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